SARCOMA OF THE TONGUE.

REPORT OF A RECENT CASE, WITH ANALYSIS OF PREVIOUSLY RECORDED CASES.

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SARCOMA of the tongue is sufficiently rare—there are, I find, only some three dozen cases on record—to make a report of an individual case justifiable, more particularly when that case presents one or two characters varying slightly from already recorded cases. I propose, therefore, to describe our own case first in some little detail, analyzing afterwards the literature upon the subject.

The patient, a man aged forty-seven years, was admitted to the Royal Victoria Hospital upon December 17, 1902, complaining of a tumor far back upon the dorsum of the tongue. He gave the history that he first noticed slight pain about the beginning of September, and that it was accompanied by a sensation as though something was stuck in the back of the throat. Only a month later did he consult a physician, who noted a mass about the size of a pea at the back of the tongue. This mass had gradually increased in size up to the time of his admission to hospital, but with this the pain had decreased, nor had there at any time been difficulty in swallowing.

Examination upon admission showed a firm, globular tumor about 1.5 centimetres in diameter situated on the right side of the dorsum of the tongue between the pillars of the fauces. It had a broad base, was sunken somewhat into the tissues of the tongue, though at the same time the outer portion rose well above the level of the surrounding parts as a rounded mass. The covering epithelium was intact and the growth extended slightly into the pharyngeal wall, but did not limit the protrusion of the tongue. There were no enlarged glands in the neighborhood, nor was anything else abnormal to be made out in the general condition of the patient.

For diagnosis the upper portion of the tumor was sliced off and removed for microscopical examination. This was hardened carefully in Zenker's fluid. Sections stained in hæmotoxylin and eosin showed that the removed portion consisted almost wholly of a cellular growth covered by a healthy epithelium. The nuclei of the cells forming this growth appeared irregularly rounded and distinctly larger than that of lymphocytes. In these first sections the cell-bodies were not sharply differentiated. The tumor appeared to consist of a mass of relatively large, rather pale-staining nuclei, embedded rather closely in a semi-translucent matrix of cytoplasm. Mitotic figures and the results of recent mitosis were to be seen frequently. No transition of these cells into forms resembling fibroblasts could be made out. Here and there a few strands of adult fibrous tissue could be recognized; in general, the vessels, which, towards the periphery, were abundant, had walls consisting of a single layer of endothelium, and these in some areas gave an obscurely alveolar appearance to the growth. This appearance was very obscure, and when present could be seen to be associated with the delicate vascular net-work, and the more the sections were studied the more they were seen to exhibit the characters of a pure sarcoma. Here and there rather diffuse collections of cells, more of the appearance of lymphocytes, could be seen.

A second portion of the growth, removed a few days later, showed more successfully the nature of the growth. In this second set of sections the individual cells were clearly defined. In general, the individual cells were found to have a relatively considerable amount of cytoplasm and to possess one nucleus, though, not infrequently, two nuclei could be seen in the one cell. The nucleus was sometimes eccentric. Now sections stained by Mallory's stain showed the arrangement of connective tissue and reticulum which is characteristic of a simple sarcoma. Broader bands of connective tissue, mostly perivascular, gave off a fine reticulum passing between the individual cells. Study of the regions where the smaller cells were accumulated led certainly to the impression that these were not simple infiltrations of leucocytes of inflammatory nature, but bore a definite relationship to the rest of the growth. As already stated, these small cells suggested lymphocytes; their nuclei were rounded and deeply staining, as compared with the more irregular, larger and more pale-

staining nuclei of the main mass of the growth. But intermingled with them, more especially towards the periphery of the masses, were what we may term cells of an intermediate order, with somewhat larger and often rather polygonal nuclei, not so deeply staining. Lastly, there were no indications of endarteritis nor of active proliferation of the connective tissue. The diagnosis was made of small round-celled sarcoma. Possibly, it would have been more correct to have spoken of this simply as a round-celled sarcoma, for the individual cells of the main mass of the growth were certainly larger than lymphocytes. I have usually classified round-celled sarcomata into the large and the small round-celled forms, and this appears to me to belong rather to the latter group than to the former. If my opinion be correct, that these larger cells have been derived from the small, deeply staining cells still present, then it would be correct to say that the growth originated as a small round-celled sarcoma.

At this period the patient was unwilling to have the operation for complete extirpation of the growth which was then recommended, and soon afterwards he entered another hospital. Unfortunately, the report of his case while there has been mislaid, so that only a very brief and probably faulty history of events can be given. So much of the tumor had been removed for examination that, with this and with possible sloughing out of the remainder, little of the tumor proper remained, or what was left became so infiltrated with inflammatory leucocytes that now the diagnosis became most doubtful. A local excision of the remaining mass was performed in January, 1903, and we learned that a section of the removed material had nothing in it to justify the diagnosis of sarcoma as distinguished from inflammatory tissue.

But about this same time the patient began to suffer from abdominal pain, and, on palpation, a mass was recognized in the epigastrium. Some weeks later an exploratory laparotomy showed the existence of an extensive growth in the region of the stomach affecting the peritoneum, portions of which, removed for diagnosis, were found to be sarcomatous. No attempt could be made to remove the growth, and, after the operation, the history was that of progressive failure of nutrition, terminating in death in August, 1903. The surgeon who attended him states that there was no sign of recurrence of the growth in the tongue. Death therefore occurred eleven months after the first onset of symptoms.

With great difficulty permission was obtained to perform a partial autopsy; examination of the abdomen alone was allowed, and permission could not be obtained either to remove or even examine the tongue. This partial autopsy was performed twelve hours after death.

Upon opening the abdomen numerous adhesions were found in the upper half; the omentum was short and thickened, a large mass lay on the posterior abdominal wall joining together the stomach, duodenum, and pancreas. These organs with the growth were removed *en masse*. And now upon naked-eye examination the growth was found to be soft, whitish in color, having in places a bluish appearance and extending through the stomach wall. This was extensively involved, and upon opening the organ a large ulcerated area was found situated about the middle of the greater curvature.

Sections of this growth showed that it was of the same nature as the original tumor in the tongue, being composed of cells of medium size with relatively large, roughly rounded nuclei, rather pale-staining, and surrounded by a fair amount of cytoplasm. These were extensively infiltrating the tissue of the part; the pre-existing fibrous stroma showing marked hyaline degeneration. Evidently, as a result of the ulceration, the tumor mass showed areas of necrosis and other areas in which there was considerable fragmentation of the nuclei, while here and there were small deposits of brownish pigment.

Obviously here was a metastasis from the original tumor, though, from the extensive nature of the growth, it is impossible to say with precision where this arose. The history renders it unlikely that the reverse was the case, and that the primary growth was abdominal.

Cases, it is true, are on record in which cancer of the tongue or œsophagus has been followed by secondary growths in the stomach, and the extensive ulceration of this latter organ in this case possibly favors to some extent the view that the metastasis began in the stomach wall. It is, however, possible that it originated in the lymph-glands behind the stomach. The point, it seems to us, must be left open, with a decision slightly in favor of gastric metastasis.

SYNOPSIS OF PUBLISHED CASES OF SARCOMA OF THE TONGUE.

INTERSTITIAL FORM.

No.	Reported by	Age and Sex.	Clinical Condition.	Histological Findings.	Treatment and Result.
I.	Santessen: Virchow-Hirsch. Jahresb., 1887, 280.	.....	A tumor of left half of tongue; gradual growth for three years; covering of mucous membranes intact; parotid metastases.	Small round cells with hyaline change in stroma.	Not treated.
II.	Butlin: Lancet, March 26, 1887, 623.	Male, 40	Pain and tumor in left middle of tongue for two years.	Lympho- or small round-celled sarcoma.	Local excision; cured; no recurrences.
III.	Targette: Guy's Hospital Reports, 1890, 21.	Male, 65	A tumor in left middle of tongue for one year, causing slight pain and limitation of movement; mucous membranes intact.	Small round-celled sarcoma.	Local removal with recurrence *in situ*. Death.
IV.	Max Sheir: Berlin klin. Woch., 1892, 534.	Male, 28	A tumor at base of tongue for about one year, causing pain and difficulty in swallowing.	Small round-celled sarcoma with infiltrating lymphocytes.	Local removal with recurrence *in situ*. Death.
V.	Denham: American Journal of Medical Sciences, 1895, 259.	Male, 61	Tumor of right side of tongue for eight months; mucous membrane covering intact.	Large round cells with reticulum.	Tongue with tumor removed; no recurrence.
VI.	Hutchinson: Medico-Chirurgical Transactions, vol. xlviii, 311.	Male, 22	A slowly growing tumor at left base of tongue for twelve years; no symptoms; mucous membranes intact.	Small round-celled sarcoma.	Total excision of tongue; recurred in floor of mouth two and a half years later. Death.
VII.	Beregzazy: Krankheiten der Zunge, 1887, 226.	Male, 42	Tumor of posterior portion of tongue for two years, causing pain and difficulty in swallowing.	Small round-celled sarcoma.	Not removed. Death. Metastases in peritoneum.
VIII.	Littlewood: British Medical Journal, February 19, 1898, 492.	Male, 17	A large tumor of middle of tongue.	Round-celled sarcoma.	Tongue and submaxillary glands removed; recurred in tonsil. Death.
IX.	Downie: British Medical Journal, October 21, 1899, 1065.	Male, 23	.....	Small round-celled sarcoma.	Tongue removed.

PEDUNCULATED FORM.

No.	Reported by	Age and Sex.	Clinical Condition.	Histological Findings.	Treatment and Result.
I.	Mikulicz u. Michelson: Atlas der Krankheiten der Mund u. Rachenhole, Heft 2.	Female, 24	A flattened pedunculated tumor on back of tongue for three months.	Not given.	Local removal.
II.	Ibid.	Male, 57	A tumor of anterior third of tongue present for six months; mucous membranes intact.	Fusiform-celled sarcoma.	Amputation of part affected; recurred *in situ*.
III.	Mercier: Rev. Méd. de la Suisse Romande, 1890.	Male, 26	A slowly growing tumor of anterior portion of tongue present for eight years; mucous membranes intact.	Large mixed-cell sarcoma.	Removed locally; no recurrence.
IV.	Targette: Guy's Hospital Reports, 1867.	Male, 2	A tumor of dorsum of tongue present for seven weeks.	Round to fusiform cells.	Removed; local recurrence. Death.
V.	Berger: Rev. de Chirurgie, 1897, 677.	Male, 26	A pedunculated tumor of left side of tongue present for six months, causing pain and difficulty in swallowing.	Fusiform-celled sarcoma.	Local excision; recurrence *in situ*. Death.
VI.	Downie: British Medical Journal, October 21, 1899, 1065.	Male, 34	A somewhat pedunculated tumor over posterior portion of tongue present for five weeks.	Spindle-celled sarcoma.	Local excision.
VII.	Melchoir-Roberts: Rev. de Chirurgie, April, 1899, 545.	Female, 64	A pedunculated tumor on right side of tongue present for three months, causing pain and fixation.	Round and fusiform cells.	Local removal; recurred *in situ*.
VIII.	Marion: Rev. de Chirurgie, 1902, 331.	Female, 37	A pedunculated tumor of left side present for four months.	Fusiform celled.	Local removal; no recurrence.

INCOMPLETE AND DOUBTFUL CASES.

No.	Reported by	Age and Sex.	Clinical Condition.	Histological Findings.	Treatment and Result.
I.	Heath: Transactions Pathological Society, London, vol. xx, 1869, 167.	Male, 60	Pain in and swelling of anterior portion of tongue for six years.	Reported as adenocarcinoma, probably a mixed-cell sarcoma.	Anterior half of tongue removed; no recurrence in nine months.
II.	Jacobi: American Journal of Obstetrics, May, 1869, 81.	Infant, 11 weeks.	A swelling of tongue noted a few hours after birth; rapid increase.	Spindle-celled sarcoma.	Left half of tongue removed. Result not stated.
III.	Huter: Berlin. klin. Woch., 1869, 346.	.....	A tumor of central posterior portion of tongue for two months.	Sarcoma.	Removed.
IV.	Albert: Wiener Med. Presse, 1885, 171.	Female, 56	A roundish tumor at root of tongue for eleven months, causing difficulty in swallowing.	Sarcoma.	Excision of tongue. Death from pneumonia in eight days.
V.	Eyre: Transactions Pathological Society, London, vol. xxxvii, 1886, 223.	.....	.....	Fibrosarcoma.	Museum specimen.
VI.	Godlee: Transactions Pathological Society, London, vol. xxviii, 1887, 346.	Female, 24	An ulcerated tumor at tip of tongue for five weeks; a calcareous nodule at centre.	Round or fusiform cells surrounding dilated cyst spaces; adenocarcinoma or mixed-celled sarcoma.	Local removal; no recurrences.
VII.	Schulten: Tinska. Lakaresalls kapets Handlingar, 1888.	Female, 32	A tumor at root of tongue, causing difficulty in swallowing.	Sarcoma.	Excision of tongue.

VIII.	Mandillon: Soc. de Med. and Clin. de Bordeaux, November 16, 1888.	Female, 21	A tumor size of a pea at tip of tongue.	Sarcoma.	Three local removals; cured.
IX.	Targette: Guy's Hospital Reports, 1873.	Male, 10 months.	A flat tumor under tip of tongue.	Sarcoma.	Local excision.
X.	Stern: Deutsch. med. Wochen., June 2, 1892.	Female, 4	A tumor of right edge of tongue present for one year.	Fusiform-celled sarcoma.	Local excision.
XI.	Onodi: Rev. de Laryngology, October 15, 1893.	Female, 17	A tumor of left base, causing limitation of protrusion with pain on swallowing; mucous membranes intact.	Elastic fibrosarcoma.	.....
XII.	Perman: Buffalo Medical and Surgical Journal, 1894, 148.	Female, 30	A tumor at base of tongue for five months, causing pain and spitting of blood.	Sarcoma.	Partial removal.
XIII.	Barker: Holmes's System of Surgery, vol. ii, 276.	.....	A pedunculated tumor of tongue with multiple sarcomata of skin.	.....	....
XIV.	Siedarovgrotzky: Jahr. v. Virchow u. Hirsch, 1873, 608.	.....	A tumor of submaxillary portion of tongue with metastases in pharyngeal glands.	Large-celled sarcoma with fatty degeneration.	.....
XV.	Poucet: Lyon. Med., 1888, 95.	Male, 32	Tumor of tongue for eight years.	Fasciculated sarcoma.	Excision of tongue; no recurrence in fifteen months.
XVI.	Eve: Transactions Pathological Society, London, 1886, 223.	.....	A tumor of left base of tongue.	Fibrosarcoma.	.....
XVII.	Bleything: New York Medical Journal, 1888, 683.	Male, 17	An ulcerated area near tip of tongue.	Sarcoma.	Cured by cauterization.

Reported cases of sarcoma of the tongue have been brought together more particularly by Sheir in 1892 and by Marion in 1897 and 1902. The total list of these writers includes thirty-five cases. Another, more recent, case has been reported by Melchoir-Roberts; but of these thirty-six several, I am inclined to think, must be set aside on the ground of imperfect diagnosis, while others are valueless on account of the imperfect nature of the reports. Excluding these, the remaining cases become grouped into two forms,—the pedunculate and the interstitial. I have in the preceding tables classified these three orders of cases, giving, I believe, the salient points of each, so far as the reports have permitted. I shall not here discuss doubtful cases nor again pedunculate cases; these latter appear almost always, if not constantly, to be composed of fusiform cells,—to be, in short, spindle-celled sarcomata.

In the other variety, the interstitial, to which our case clearly belongs, there are several in which the details approximate closely to those here given of our own case.

Thus, Hutchinson's case, reported in 1885 in the *Lancet*, was that of a man, aged twenty-two, having a tumor upon the dorsum of the tongue in the posterior portion of the left half which had exhibited a steady, slow growth for twelve years. There were no enlarged glands, nor was there any fixation of the organ. The tongue was removed, but the growth recurred *in situ* two and a half years later, followed by death in a short period. The pathological diagnosis was small round-celled or lymphosarcoma.

Santesson reports a somewhat similar case in 1887. The tumor was in the left half of the tongue and had been growing for three years. There were metastases in the neighboring glands. The growth consisted of small round cells having a fibrous tissue reticulum which showed marked hyaline degeneration.

Butlin reported another case in the same year, that of a man of forty years who had suffered from a painful mass at the left side of the tongue. There was a single enlarged gland in the neighborhood; the covering epithelium was intact, and iodides were ineffectual. Here, also, the pathological diagnosis was that of lymphosarcoma, or small round-celled sarcoma. It is to be noted that after local excision there was no recurrence.

A third case, also in 1887, was that of Beregzazy. Here, too, the case was that of a man, aged forty-two, having a steadily growing tumor in the posterior portion of the tongue for about two years, with no difficulty in swallowing. There is no history given of local metastasis nor of

operation, but, death occurring, metastases were found in the peritoneum, most extensive at the attachment of the mesentery to the bowel, where small semilunar masses were present. Again the microscopical diagnosis was small round-celled or lymphosarcoma.

Sheir's case in 1892 was of the same order, though his patient was younger. A man aged twenty-eight had suffered for two years from a tumor at the base of the tongue, which had attained almost the size of an egg, and caused pain and difficulty in swallowing. It was removed through a submaxillary incision when it shelled out. There was an apparent capsule which also was removed. The growth, however, soon recurred in the floor of the mouth, leading to death fifteen months later. The microscopical diagnosis in this case was that of a small round-celled sarcoma; and it was noted in the report that besides the proper tumor cells there were collections or groups of cells exactly resembling lymphocytes.

Analyzing these cases, it will be seen that they all belong to the group of round-celled sarcomata; that they occur, not in early life, but in middle age, the majority between the ages of forty and fifty, though one occurred as early as twenty-eight; that extensive metastases in the neighborhood are rather the exception than the rule, and that other cases besides ours have shown lack of such local metastasis; that, as a body, they differ from the ordinary round-celled sarcoma in their relatively slow growth. Though local recurrence may take place, still, in Butlin's case and ours, it would seem to have been wanting. In Bergzazy's case, as in ours, though there was no local recurrence, metastases were found in the abdomen.

The clinical diagnosis of these tumors presents some little difficulty. They are distinguished from cancerous growth proper by the fact that the epithelium usually remains intact or only ulcerates after a long period. From gummata they ought to be distinguished by the therapeutic test. It is not always easy under the microscope to differentiate a gummatous growth from small-celled sarcoma. That is, there are portions of a gummatous growth which, from the abundance of small round cells, may be mistaken for a malignant connective-tissue neoplasm. But where the whole growth consists of the one order of cells with a mesh-work of well-developed connective tissue, there can be no doubt that we are dealing with a sarcoma proper.

Coming now to the different forms of sarcoma, we are immediately met with a difficulty regarding the terminology. It will be seen from what I have stated that the majority of these cases have been regarded as possibly lymphosarcoma. This term is a bugbear to the modern pathologist, for it is employed by different individuals with different meanings. Thus, many American authorities appear to confine the term largely to the overgrowth of lymph-glandular tissue in pseudolcukæmia. French writers employ the term, it would seem, for any small round-celled sarcoma that shows an intracellular reticulum at all resembling that seen in true lymphoid tissue, while many English writers employ it as synonymous with small round-celled sarcoma. Nor do I feel wholly convinced that it is possible to make an absolute distinction between the small round-celled sarcoma and the lymphosarcoma, for, stained with one of the connective-tissue stains, such as Mallory's, undoubtedly the true small-celled sarcoma does possess an intracellular reticulum. It would be better if, under these circumstances, the use of this term were wholly done away with until an absolute histological differentiation is afforded between the two varieties, if such truly exist, or, with the German authorities, to apply the term purely to those cases where there can be no question of the growth having originated in true lymphoid tissue. I would only add that, while the growth in our case cannot be described as one of the smallest of the round-celled sarcomata,—as a lymphocytoma, if I may coin the word,—the relationship of the small deeply staining cells present to the other cells would appear to indicate that this case of ours falls well into line with the others described as small round-celled (or lympho-) sarcoma.

Lastly, as to treatment. That most in favor is wide excision of the tumor. This may result in cure. While there may be recurrence, either local or distant, the results here given seem to justify that this be recommended. The slow growth of many of these tumors would also seem to suggest that if excision be done at an early date, the results should be most favorable. In our case, owing to the opposition of the patient,

only partial excision was performed; but this partial excision, while it led to the arrest of the growth locally, may, I would suggest, have been not unfavorable to the escape of the tumor cells and to the establishment of distant metastases.

In conclusion, I wish to express my thanks to Professor Adami for advice and in the preparation of the foregoing.